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CASE REPORT

Charles Bonnet syndrome in cranio-maxillofacial surgery: case report

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Abstract
Background Complex visual hallucinations in the presence of a clear mental state and in the absence of underlying neurological disorders have received increased recognition since Charles Bonnet reported visual hallucinations secondary to mature cataracts in 1760. The prevalence of Charles Bonnet syndrome (CBS) varies widely in the literature and might be underestimated in most settings. The current paper presents a case of acute-onset CBS due to a unilateral Frost suture after revision of an orbit floor fracture.
Case report A 68-year-old male patient underwent an operation to repair a unilateral orbital floor fracture and subsequent eye patching by a Frost suture. The patient complained of complex, colored visual hallucinations ~3 h after waking from general anesthesia. The visual hallucinations stopped during sleep and reappeared in the morning. The symptoms disappeared completely ~2 h after removal of the Frost suture.
Discussion Frost sutures are commonly used in oculoplastic surgery and may result in acute onset of visual hallucinations. CBS is often neglected, and clinicians must be aware of the association between acute visual deprivation and CBS.

Keywords Frost suture · Charles Bonnet syndrome · Hallucinations

Background

Charles Bonnet, a Swiss philosopher and naturalist, described a phenomenon of complex visual hallucinations in his grandfather in 1760. The phenomenon occurred due to visual impairment because of bilateral cataracts in the absence of underlying neurological or psychological disorders [1, 2]. The syndrome was named after Charles Bonnet in 1937 by de Mosier [2]. Charles Bonnet became severely visually handicapped due to vision loss of unknown cause, experiencing complex visual hallucinations [1]. In 1902, Fluornoy wrote an essay in which he described personal visual experiences of Bonnet's grandfather, which he dictated to his grandchild more than a hundred years before.

Hallucinations are defined as visual, auditory, or sensory perceptions in the absence of an external stimulus. In Charles Bonnet syndrome (CBS), the hallucinations are usually of a complex visual nature, but auditory hallucinations may also occur [3, 4]. Schadlu et al. described risk factors for CBS as follows: age of >64 years, social isolation, lower cognitive function, history of stroke, poor lighting, and impaired bilateral visual acuity [3, 5]. Nevertheless, CBS may occur in any patient suffering from visual impairment. The hallucinations usually start with visual impairment; however, they may occur after a delay, and their frequency can vary [3, 5–9]. Patients commonly describe their hallucinations as non-disturbing, and they have insight into the unreality of the hallucinations [3, 4].

The pathogenesis of CBS has not been fully understood until now. Two theories may explain the genesis of CBS. The "release theory" ascribes hallucinations to a lesion of the visual pathway resulting in abnormal signals being sent to the visual cortex, whereas the "deprivation theory" suggests that impairment of sensory input leads to formation of spontaneous images in the visual association cortex [3, 4, 6].

The mainstay of treatment in patients suffering from CBS is improvement of visual acuity and an empathetic understanding that the patients' symptoms are not forerunners of a psychiatric disorder [3, 7]. Furthermore, different pharmacological agents are applied to treat hallucinations associated with CBS; for example, atypical neuroleptica, selective serotonin reuptake inhibitors, or reversible cholinesterase inhibitors [10–12].

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Visual impairment is a common side effect in patients with cranio-maxillofacial injury caused by trauma, as is early postoperative swelling. However, CBS is not a frequently described side effect in these patients. To the best of the author's knowledge, no cranio-maxillofacial journal has described this topic; information on CBS usually occurs in ophthalmologic or psychiatric literature.

The present paper describes a case of acute-onset CBS due to visual deprivation after application of a Frost suture.

Case report

Patient

A 68-year-old male patient was referred to our department with a diagnosis of a dislocated left orbital floor fracture due to a syncopal event at home 12 h previously. Initial computed tomography, performed in an outsource hospital, showed no further pathologies, and particularly no intracranial bleeding. The patient presented with a Glasgow coma score of 15, a periorbital hematoma on the left side, and double vision during the forced upward glance. The ophthalmologic examination on arrival showed no impairment of visual acuity, and there was no indication of incarceration of the inferior rectus muscle.

In terms of concomitant diseases, the patient suffered from arterial hypertension, prostate hypertrophy, extreme obesity (body mass index, 41 kg/m^2), and chronic obstructive pulmonary disease. Antihypertensive therapy comprised combined treatment with spironolactone (Aldactone®; Pfizer Schweiz AG, Zürich); aliskiren, amlodipine, and hydrochlorothiazide (RasilAmlo®; Novarits Pharma Schweiz AG, Bern); and bisoprolol (Bilol®; Sandoz Pharmaceuticals Schweiz AG, Steinhausen). For treatment of prostate hypertrophy, therapy with sabalis serrulatae fructus (Prostagutt-F®; Schwabe Pharma Schweiz AG, Küssnacht am Rigi) had been initiated by his family doctor, who also began prophylactic administration of acidum acetylsalicylicum (Aspirin Cardio®; Bayer Schweiz AG, Zürich) about 2 years previously. The patient had himself discontinued continuous positive airway pressure therapy 5 years previously.

Three days after the trauma, surgical revision of the orbital floor was performed by a transconjunctival approach, and titanium mesh was inserted and adapted to the extent of the defect. In addition, for prophylaxis of entropion and ectropion, a Frost suture (Fig. 1) was applied.

About 3 h after waking from general anesthesia, the patient reported complex visual hallucinations, such as shaped figures creeping across the room. He was unable to characterize the hallucinations more precisely. Although the patient was at all times aware that these images were not real, he felt nervous and insecure. Neurological examination at the time of presentation was inconspicuous. The Frost suture was removed immediately because diagnosis of CBS was assumed. Ophthalmological examination after removal showed an uncorrected visual acuity of 0.6 in the affected and 0.3 in the contralateral eye. The hallucinations showed complete remission 2 h after removal of the Frost suture, and no relapse was observed.

Fig. 1 Frost suture in place and taped to the supraorbital skin

Discussion

In this manuscript, a male patient with iatrogenic CBS after surgical therapy of an orbital floor fracture and postoperative "taping" with a Frost suture is presented. The complex visual hallucinations were ascribed to the Frost suture because his medication did not predispose to hallucinations and the operative trauma was unlikely to cause these kinds of hallucinations.

The Frost suture, as initially described by Albert Frost in 1934, functions to support the upper eyelid after ptosis correction [13]. To avoid lower eyelid retraction due to scarring or over-resection of the lower lid skin, resulting in ectropion or entropion, a modification of the original technique has been proposed by several authors [14–16]. Frost sutures are applied after a transconjunctival approach in a proportion of patients with orbital wall fractures with pronounced pre- and intraoperative swelling. In these patients, the Frost suture is placed through the gray line of the lower lid using a resorbable suture (Vicryl Rapid 5-0, Ethicon, Norderstedt, Germany). To support the lower lid, the two ends of the suture are taped to the skin of the supraorbital region (Fig. 1).

CBS is usually described in context with a slowly progressive loss of visual acuity. Few patients with acute-onset CBS have been described in the literature; indeed, this is only the third report of CBS following eye patching [17–19]. Other causes of CBS following acute visual loss, such as unilateral enucleation, macular translocation surgery, multiple sclerosis, or central retinal artery occlusion, have been described by other authors (Table 1) [20–25].

Table 1 Descriptive statistics of patients with CBS in the literature including the described case, subdivided in type of visual impairment

Descriptive statistics	Eye patching [17–19]	Enucleation [22]	Multiple Sclerosis acute attack [23]	Macular translocation surgery [20, 21]	Central artery occlusion [25]
n patients (total *n*=9)	3	1	1	2	2
Female (total *n*=5)	1	0	1	2	1
Male (total *n*=4)	2	1	0	0	1
Average age (years) (of all patients 73 years)	73	65	–	83.5	70
Range (years)	68–80	–	–	83–84	63–77
Average timespan 1 (h)[a]	17	2160	2,928	24	96
Range (h)	0.17–48	–	–	24	48–144
Average timespan 2 (h)[b]	33	No remission	–	120	1,428
Range (h)	2–48	No remission	–	72–168	3–14 (weeks)

[a] Average timespan 1: timespan from the start of visual impairment until the onset of hallucinations

[b] Average timespan 2: timespan from the start of CBS until complete remission

–No information available

The delay between visual loss and onset of complex visual hallucinations in these cases varies from 10 mins to 4 months, whereas the average timespan from visual impairment and onset of hallucinations in patients with eye patching, macular translocation surgery, and central artery occlusion ranges from 17 to 96 h. The average timespan in patients suffering from attacks of multiple sclerosis or enucleation ranges from 3 to 4 months. Acute and non-acute onsets are not clearly defined in the literature. Patients describe typical CBS-associated hallucinations such as persons, colored objects, and animals moving in the room, and the patients maintain their orientation and consciousness during the hallucinations.

As previously mentioned, the pathophysiology of CBS is not fully understood. Two hypotheses have been proposed, of which the "deprivation theory" may explain the origin of the present case of CBS [3, 4]. The early onset and the immediate, complete remission of the visual hallucinations after removal of the Frost suture together with a lack of disorientation supports this theory.

The hallucinations associated with CBS can vary widely. Even auditory hallucinations have been reported; however, their coexistence is controversial [3, 4]. The precise description of the shape or position of persons, objects, and animals is suggestive of CBS-associated visual hallucinations. The hallucinations cannot be explained by the presence of a psychiatric disorder and they are usually not disturbing, although some patients have reported tension and anxiety during the hallucinations [3, 5, 7]. There is typically insight into the unreality of the hallucinations; however, patients may initially feel confused at the appearance of the images [3, 5, 7]. Lack of insight should lead to consideration of differential diagnoses including conditions with psychotic symptoms such as bipolar disorders, Alzheimer's disease, or Parkinson's disease. In addition, delirium, drug intoxication, drug withdrawal, or adverse effects of medications may lead to visual hallucinations. Although most patients with CBS do not suffer from underlying psychiatric disorders, dementia due to Lewy body disease could be a coexisting diagnosis, especially in elderly patients [5].

The prevalence of CBS is 10–15 % among visually impaired patients, which might be a gross underestimation because of the high rate of patients who do not present to hospital due to the fear that their hallucinations may be labeled as part of a psychological disorder. Vukicevic et al. performed a multicenter screening of 200 patients aged 60 years and older using a specific questionnaire [8]. They showed that 21 % of patients with CBS did not report their symptoms to anyone, 64 % mentioned them to a family member, and only 15 % reported their hallucinations to their care provider. This study suggests the importance of dialogue with the patient as well as family members and specialists [3, 5, 8, 9].

To avoid or diminish the risk of postoperative scarring leading to entropion or ectropion, modified Frost sutures are applied in a few special cases with pronounced pre- or intraoperative swelling during oculoplastic surgery such as blepharoplasty or transconjuctival approaches to the orbital floor. With elevation of the upper eyelid, vision can be checked and eye drops can easily be applied if necessary. To the author's knowledge, no prospective or retrospective study of Frost sutures avoiding or diminishing the risk of lower lid retraction has been performed. More research on the effect of Frost sutures should be performed [13–16].

Covering the eye is a common procedure in craniomaxillofacial surgery, ophthalmology, and other specialties. As CBS becomes more prevalent in the aging population, clinicians who care for elderly patients must be aware of the

characteristics of and treatment options for CBS and consider that CBS may occur in association with temporary eye patching [5]. Patients should be informed of the risk of hallucinations and be reassured of their benign and usually temporary nature. Removal of the eye patch and an empathetic understanding of the patient are key steps in treatment of CBS in these cases. Interdisciplinary pharmacological treatment should be discussed in cases of persistent complex visual hallucinations [10–12].

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